Mastery Motivation in Children with Intellectual Disability: Is There Evidence for a Down Syndrome Behavioural Phenotype?

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Abstract

The main purpose of the current study was to provide empirical evidence to support or refute assumptions of phenotypic deficits in motivation for children with Down syndrome. Children with moderate intellectual disability associated with etiologies other than Down syndrome were recruited in an extension of a previous study that involved children with Down syndrome and typically developing children. The participants were 29 children with moderate intellectual disability and 33 children with Down syndrome who were matched on mental age to 33 typically developing children, aged 3 to 8 years. Mastery motivation was assessed on task measures of curiosity, preference for challenge, and persistence, as well as parental reports. There were no significant group differences on the mastery motivation tasks. Parental ratings of mastery motivation differed, with typically developing children generally being rated more highly than each of the disability groups. The view that motivational deficits are part of the Down syndrome behavioural phenotype was not supported.

Keywords: motivation, mastery motivation, intellectual disability, Down syndrome, behavioural phenotype, persistence

Motivation in Adolescents with Intellectual Disability: Is There Evidence for a Down Syndrome Behavioural Phenotype?

It has been suggested that motivational deficits are part of the learning and behavioural phenotype of Down syndrome. Fidler (2006) proposed that primary phenotypic deficits in cognition combine with primary strengths in social functioning to produce a secondary phenotypic pattern of motivation that is characterised by low levels of engagement and persistence. Evidence of differences in motivation between individuals with Down syndrome and those with intellectual disability associated with other etiologies is essential for establishing phenotypic profiles of behaviour (Dykens. 1995) but such differences have not yet been documented.

The overwhelming majority of motivation studies involving children with Down syndrome have involved comparisons with groups who are developing typically, usually matched for mental age, and with a focus on the early childhood years. Some differences in approaches to learning have been identified. Wishart and her colleagues (Duffy & Wishart, 1987; Pitcairn & Wishart, 1994; Wishart, 1991) reported task avoidance, counterproductive behaviours and unstable performance in young children with Down syndrome, leading them to conclude that the development of motivation is not only delayed but also "fundamentally different" (Wishart, 1999, p. 497). In addition, the tendency for children with Down syndrome to seek social interactions with others was hypothesised to interfere with their independent mastery attempts (Pitcairn & Wishart, 1994).

Most work in the area of motivation of individuals with intellectual disability has used the paradigm of mastery motivation which conceptualises motivation as a striving to achieve competence (Morgan, Harmon, & Maslin-Cole, 1990; Yarrow et al., 1983). There are two main ways of measuring mastery motivation: laboratory tasks and parent or teacher questionnaires. These methods have produced contrasting results. When tasks to measure

behaviours such as persistence and preference for challenge are used, few differences have been demonstrated between children with Down syndrome and typically developing children of the same mental age (Gilmore, Cuskelly, & Hayes, 2003; Glenn, Dayus, Cunningham, & Horgan, 2001; MacTurk, Vietze, McCarthy, McQuiston, & Yarrow, 1985) although finer grained analyses sometimes reveal subtle differences (Ruskin, Mundy, Kasari, & Sigman, 1994). By contrast, children with Down syndrome have been consistently rated by their parents as lower in mastery motivation than typically developing children (Gilmore et al., 2003; Glenn et al., 2001; Ruskin et al., 1994). Although it is possible that parents' observations are a better reflection of a child's everyday motivation for mastery, it is likely that their ratings are influenced by chronological-age expectations, unlike laboratory tasks which involve comparisons on the basis of mental age (Cuskelly, Gilmore, & Carroll, 2013).

In a recent study, we investigated motivation in 10-15 year old children with Down syndrome and a comparison group of typically developing children matched for mental age (3 to 8 years) (Gilmore & Cuskelly, 2011). As found in most of the previous work with younger children, there were no group differences on task measures of persistence, curiosity and preference for challenge. One persistence task did approach significance, however, with a moderate effect size. Typically developing children displayed greater persistence, and we speculated that, in a larger sample, this difference may have reached significance. Consistent with earlier research, parents of typically developing children reported them to be significantly more persistent compared to the children with Down syndrome.

Difficulties with motivation, if they exist, may potentially be attributable to a range of factors associated with intellectual disability, and to the various experiences and opportunities that are available. But these difficulties cannot be considered part of the behavioural phenotype without robust evidence of differences between individuals with Down syndrome and those whose intellectual disability is due to other causes. If motivation deficits are

indeed part of the Down syndrome behavioural phenotype, we would expect to see significant group differences on measures of motivation.

The main purpose of the current study was to provide empirical evidence to support or refute assumptions of phenotypic deficits in motivation for children with Down syndrome. To achieve this aim, we added a sample of children with moderate intellectual disability due to causes other than Down syndrome to our previous study of children with Down syndrome and typical development (Gilmore & Cuskelly, 2011). We retained the typically developing group in our analyses in order to test the possibility of motivational deficits associated with intellectual disability per se. There is little empirical evidence to support this possibility, other than questionnaire data from parent reports (Zigler, Bennett-Gates, Hodapp, & Henrich, 2002) and a study conducted more than 40 years ago (Harter & Zigler, 1974). Nevertheless, the assumption that low motivation is inherent to intellectual disability appears to be quite pervasive (see, for example, Bennett-Gates & Zigler, 1999; Deci, 2003; Greenspan, 2006; Merighi, Edison, & Zigler, 1990; Switzky, 1997).

Method

Participants

The participants were 29 children with moderate intellectual disability, aged from 8 to 16 years who were combined with the 33 children with Down syndrome (aged 10 to 15 years) and 33 typically developing children (aged 3 to 8) reported by Gilmore and Cuskelly (2011). The sample with moderate intellectual disability was selected to match on mental age (MA) with the group with Down syndrome, which had previously been matched to chronological age (CA) of the typically developing group. A one-way analysis of variance revealed no significant CA-MA differences. Using Fisher's exact test, there were no significant differences on maternal or paternal education across the three groups. Descriptions of the sample are shown in Table 1. Within the moderate intellectual disability group, there was a

range of organic etiologies including Noonan's syndrome, Fragile X, Kabuki syndrome, and William syndrome.

INSERT TABLE 1 ABOUT HERE

Measures

Children completed the motivation tasks reported in Gilmore and Cuskelly (2011). These included the curiosity and preference for challenge measures originally used by Harter and Zigler (1974) as well as two additional tasks created by Gilmore and Cuskelly (2011) to assess persistence.

Curiosity. The measure of curiosity requires the child to choose between two doors on a series of wooden houses. One of each pair of doors has a picture on the front and the child is told that the same picture is located behind this door. The second door is blank and the child is told that behind this door there is a new picture, one the child has not seen before. After a demonstration and a sample item, the child is presented with 10 separate trials using different houses. The curiosity score ranges from 0 to 10 based on the number of trials on which the child chooses to open the blank door.

Preference for Challenge. Preference for challenge is measured on a series of three sets of puzzles, each of which comprises three identical 15 or 16 piece puzzles. Each set is presented with varying numbers of pieces removed from the three puzzles to produce three levels. The easy version has five pieces needing to be replaced, the medium level has 10 pieces to insert, and the difficult one has all but two pieces removed. Children are asked to choose one puzzle to complete, and their choice is scored 1 = easy, 2 = medium or 3 = difficult. Thus, the maximum obtainable score for the measure is 9, with higher scores representing a greater preference for challenge.

Persistence. The two persistence tasks were designed so that children are able to achieve some success but cannot complete the entire task. The picture search task requires the

child to locate seven specific images on a large sheet of approximately 250 small pictures of objects, animals, and figures (copied, with permission, from pages 30-31 in Wick & Marzollo, 1995). Two of the seven images are not present on the sheet, making this an impossible task. During a 10 minute period, the examiner calculates the number of 15 second intervals during which the child is predominantly task focused, producing a score between 0 and 40. The standard procedures designed by Morgan, Busch-Rossnagel, Maslin-Cole and Harmon (1992) are followed for prompts and termination.

The fishing task requires the child to use a magnetic fishing rod to attract 10 magnetic sea creatures out of a bowl. Some are easier to pick up than others due to the varying strengths of the magnets embedded in the creatures, and a few are impossible. The same scoring procedures are used as for the picture search task, with children being given one point for every 15 second interval of task engagement.

Dimensions of Mastery Questionnaire (DMQ-17*;* Morgan, Leech, Barrett, Busch-Rossnagel, & Harmon, 2002). The DMQ-17 assesses parental perceptions of children's motivation. The measure comprises 45 items that are rated on a 5-point scale from 'not at all typical' to 'very typical'. There are four scales reflecting the instrumental aspects of mastery motivation: Object Persistence, Gross Motor persistence, Social Persistence with Adults, and Social Persistence with Children, as well as two scales that assess expressive aspects (Mastery Pleasure and Negative Reaction to Failure). In the current study, subscale reliabilities were adequate to good for most scales (.69 to .93). In two of the three groups, Cronbach's alphas for Negative Reaction to Failure were unacceptably low (.59 in the Down syndrome group and .49 in the moderate intellectual disability group); thus, this subscale was omitted from further analyses.

Procedure

Children with moderate intellectual disability were recruited from mainstream and special schools. Sessions were conducted by the third author in a private room at the children's schools, beginning with administration of the Stanford Binet 4th edition which provided a measure of mental age. Consistent with the procedures used previously with the Down syndrome and typically developing groups (Gilmore & Cuskelly, 2011), the motivation tasks were then administered in the following order: curiosity, preference for challenge, and the two persistence tasks which were counterbalanced. For all tasks, the standard procedures with respect to administration (i.e., instructions, prompts and feedback) were followed as detailed by the researchers (Harter & Zigler, 1974; Morgan et al., 1992). The DMQ was sent home for parents to complete, and all but five families in the moderate intellectual disability group returned the questionnaire.

Results

Statistical analysis was performed using SPSS 19. Prior to analysis, the data were screened and no significant breaches of normality were identified in the distributions of scores. A one-way analysis of variance (ANOVA) was conducted for each motivation task. There were no significant group differences on any of the measures. Means and standard deviations are shown in Table 2. To compare performance of the two disability groups more directly, independent samples *t*-tests were conducted. The results were not significant, and the highest effect sizes were relatively small (d = .34 and .35, respectively, for the picture search and fishing tasks).

INSERT TABLE 2 ABOUT HERE

Using the five internally consistent subscales of the DMQ, a multivariate analysis of variance (MANOVA) was significant, $F(5, 84) = 7.67$, $p < .001$, partial $\eta^2 = .31$. As shown in Table 3, all subscales, with the exception of Mastery Pleasure, differed significantly between groups. Post hoc comparisons using Tukey HSD showed that typically developing

children were rated more highly on Object Persistence, Gross Motor Persistence and Social Persistence with Children than were the two disability groups. They also obtained higher scores for Social Persistence with Adults than the group with moderate intellectual disability. There were no significant differences in parent ratings for the two disability groups.

INSERT TABLE 3 ABOUT HERE

Correlations among the measures for each of the three groups are shown in Tables 4, 5 and 6. For the task measures, there were significant relationships between the two persistence tasks (picture search and fishing) in all three groups. Only in the group of children with moderate intellectual disability were there any significant correlations for the other tasks. Curiosity and preference for challenge were related, as were preference for challenge and persistence with picture search.

INSERT TABLE 4 ABOUT HERE

INSERT TABLE 5 ABOUT HERE

INSERT TABLE 6 ABOUT HERE

There were significant relationships among most DMQ subscales, particularly in the typically developing group. Some correlations were lower and non-significant in the two disability groups. The only significant relationship between task measures and parent report in the moderate intellectual disability group was a negative relationship between task curiosity and parent-reported object persistence. In the Down syndrome group, persistence on the picture search task was significantly related to parent reported object persistence and negatively related to social persistence with adults. Preference for challenge was significantly related to parental reports of both object and gross motor persistence for typically developing children, and there was a significant negative relationship between curiosity and social persistence with children.

Discussion

Understanding phenotypic characteristics has the potential to guide intervention efforts, as well as to contribute to understanding brain-behaviour relationships (Hodapp & Fidler, 1999; Reilly, 2012). A key issue not previously addressed in motivation research is the extent to which children with Down syndrome differ from those with intellectual impairments associated with other etiologies. The results from our investigation show no significant group differences on the laboratory tasks measuring mastery motivation, and the expected differences on parent report between the disability groups and the typically developing group.

Previously, we showed that children with Down syndrome performed similarly to typically developing children on motivation measures of curiosity, preference for challenge, and persistence (Gilmore & Cuskelly, 2011). Scores for the new sample of children with moderate intellectual disability show no significant differences from the other two groups. In the earlier study, we noted one motivation measure that was worthy of future investigation. Although not quite reaching the level set for significance, persistence on the picture search task had a moderate effect size. Findings from the current study show that scores on this task for the group with moderate intellectual are more similar to those of the typically developing group. The difference between the two disability groups is not significant, however, and the effect size is relatively small.

The results from parent ratings are consistent with earlier work. Parents of typically developing children rated them as significantly more persistent with objects, tasks requiring motor skills, and social interactions with other children. Interestingly, children with moderate intellectual disability were rated as less socially persistent with adults than were typically developing children. This difference is not evident for the group with Down syndrome, a finding that is in line with the view that social competence is a relative strength for children

with Down syndrome (Rosner, Hodapp, Fidler, Sagun, & Dykens, 2004; Sigman & Ruskin, 1999).

As mentioned earlier, one potential reason for the differences between mastery motivation scores obtained from laboratory tasks and parent report is that, when completing the questionnaire, parents may be comparing their child with others of the same chronological age, without taking into account their child's developmental delays. Future research could explore this possibility by modifying the instructions given to parents before they complete the DMQ, or by questioning them later to identify their frame of reference when responding. As well, the possibility remains that the laboratory measures, being single snapshots of performance on a particular day, do not capture the child's mastery motivation adequately. Parents who have observed their child's performance across time and multiple settings may in fact provide a more accurate picture of their general motivation; on the other hand, some of their observations will occur when the child is faced with tasks that are either too easy or too difficult, conditions under which mastery motivation cannot be demonstrated. The structured nature of the laboratory tasks and their method of administration may also be relevant. It is possible that children with intellectual disability are less able to maintain their engagement with activities that have lower levels of structure, even if they meet the criterion of optimal challenge.

There are some limitations that should be kept in mind when interpreting the results, including our relatively small sample size and the difficulties of interpreting differences in mastery motivation scores derived from laboratory measures and parent report. Like most previous studies, the samples were formed by matching mental age of the two disability groups to chronological age of the typically developing group, and the samples were thus notably different with respect to their years of schooling and other life experiences. As we have argued elsewhere, similar mastery motivation task scores may be the product of

different processes (Gilmore et al., 2003). In particular, the fact that the disability and typically developing groups differed markedly on chronological age is relevant, since experience is generally understood to play an important role in mastery motivation (Cuskelly & Gilmore, 2014).

Conclusions

The results from our expanded study of mastery motivation suggest that children with Down syndrome do not differ significantly from others with similar levels of intellectual disability. Although we need to be cautious in our conclusions because of the study's relatively small sample size, the view that motivational deficits are part of the Down syndrome behavioural phenotype is not supported by the evidence presented here. The two disability groups are distinguished only by the finding that children in the moderate intellectual group are less persistent in their interactions with adults than are typically developing children, a difference that is not evident for children with Down syndrome. Further research with larger samples is needed. Comparisons with other specific syndrome groups (e.g., Williams or Prader-Willi) would be preferable, given the potential confounds that may be present in a mixed etiology group.

Our second conclusion is that the findings from the task measures provide no evidence to support assumptions about a link between motivational deficits and intellectual disability. These assumptions need to be challenged in larger scale research. It is possible that other characteristics associated with intellectual disability (e.g., slower processing speed or passivity), are sometimes interpreted as motivational (Goodman and Linn, 2003). It would be important to attempt to identify such misinterpretations as they are likely to influence attributions and expectations by others, potentially reducing the opportunities that individuals with intellectual disability have to achieve mastery and competence.

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Table 1

Chronological Age (CA) and Mental Age (MA) of the Sample

	Down syndrome (n = 33) 16 females		Moderate ID (n = 29) 14 females		Typically developing (n = 33) 21 females	
	Mean	*(SD)*	*Mean*	*(SD)*	*Mean*	*(SD)*
CA (months)	157.06	(18.36)	146.83	(23.56)	60.18	(16.01)
MA (months)	54.58	(13.50)	55.86	(11.47)		

Table 2

Comparisons of Children with Down Syndrome (DS), Moderate Intellectual Disability (MID), and Typical Development (TD) on the Four Mastery Motivation Tasks

Measure	Possible score range	DS (n = 33) Mean (SD)	MID (n = 29) Mean (SD)	TD (n = 33) Mean (SD)	*F* (df = 2,92)	*p*	Partial η^2
Curiosity	0 – 10	4.91 (3.30)	5.66 (3.02)	5.45 (3.61)	.43	.66	.009
Preference for Challenge	3 – 9	5.45 (1.97)	5.55 (1.90)	5.54 (2.08)	.02	.98	.001
Persistence: Picture Search	0 – 40	16.70 (11.74)	20.79 (12.42)	21.73 (10.38)	1.77	.18	.037
Persistence: Fishing	0 – 40	25.06 (11.19)	29.10 (11.72)	27.06 (10.76)	1.01	.37	.021

Table 3

Comparisons of Children with Down Syndrome (DS), Moderate Intellectual Disability (MID), and Typical Development (TD) on the Dimensions of Mastery Questionnaire (DMQ)

Subscale	DS Mean (SD) n = 33	MID Mean (SD) n = 24	TD Mean (SD) n = 33	*F* df = 2,87	Partial η^2
Object Persistence	2.62 (0.92)	2.59 (0.88)	3.49 (0.46)	13.92 ***	.24
Gross Motor Persistence	3.02 (0.98)	3.03 (0.88)	3.75 (0.80)	7.01 **	.14
Social Persistence Adults	3.80 (0.69)	3.60 (0.75)	4.16 (0.53)	5.42 **	.11
Social Persistence Children	3.42 (0.92)	3.42 (0.97)	4.30 (0.64)	11.43 ***	.21
Mastery Pleasure	4.33 (0.63)	4.13 (0.63)	4.47 (0.48)	2.43	.05

** $p < .01$, *** $p < .001$

Table 4
Pearson Correlations (one-tailed) of all Measures for DS group (n = 33)

Measure	CUR	PC	PS	FI	OP	GM	SPA	SPC
Task Curiosity (CUR)	1							
Task Preference for Challenge (PC)	.18	1						
Task Picture Search Persistence (PS)	.18	-.14	1					
Task Fishing Persistence (FI)	.27	.02	.38*	1				
DMQ Object Persistence (OP)	.04	-.08	.48**	.29	1			
DMQ Gross Motor Persistence (GM)	.08	-.13	.00	.11	.53***	1		
DMQ Social Persistence Adults (SPA)	-.12	.08	-.53***	-.11	-.05	.28	1	
DMQ Social Persistence Child (SPC)	.21	-.04	-.17	.08	.32*	.53***	.45**	1
DMQ Mastery Pleasure (MP)	-.09	-.07	-.09	-.21	.11	.13	.34*	.36*

* $p < .05$, ** $p < .01$, *** $p < .001$

Table 5
Pearson Correlations (one-tailed) of all Measures for MID group (n = 29 for task measures; n = 24 for DMQ measures)

Measure	CUR	PC	PS	FI	OP	GM	SPA	SPC
Task Curiosity (CUR)	1							
Task Preference for Challenge (PC)	.47**	1						
Task Picture Search Persistence (PS)	.18	.43*	1					
Task Fishing Persistence (FI)	.08	.25	.62**	1				
DMQ Object Persistence (OP)	-.35*	.09	.27	.09	1			
DMQ Gross Motor Persistence (GM)	-.08	-.14	.18	.12	.69**	1		
DMQ Social Persistence Adults (SPA)	-.14	.27	.07	.00	.34	.44*	1	
DMQ Social Persistence Child (SPC)	-.20	.06	-.05	.02	.26	.52**	.76**	1
DMQ Mastery Pleasure (MP)	.11	.10	.18	-.01	.12	.33	.65**	.45*

* $p < .05$, ** $p < .01$, *** $p < .001$

Table 6
Pearson Correlations (one-tailed) of all Measures for TD group (n = 33)

Measure	CUR	PC	PS	FI	OP	GM	SPA	SPC
Task Curiosity (CUR)	1							
Task Preference for Challenge (PC)	.14	1						
Task Picture Search Persistence (PS)	-.04	-.02	1					
Task Fishing Persistence (FI)	.13	.12	.42**	1				
DMQ Object Persistence (OP)	.18	.41**	-.18	.23	1			
DMQ Gross Motor Persistence (GM)	-.11	.30*	.07	.17	.42**	1		
DMQ Social Persistence Adults (SPA)	-.06	.21	.04	.01	.38*	.42**	1	
DMQ Social Persistence Child (SPC)	-.37*	.27	.12	.12	.37*	.50**	.59***	1
DMQ Mastery Pleasure (MP)	.09	.25	.07	.07	.32*	.05	.60***	.40*

* $p < .05$, ** $p < .01$, *** $p < .001$